CASE REPORT

An Unusual Presentation of Parathyroid Adenoma: A Radiological Point of View

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ABSTRACT

Bone tumors are characterized by hyperparathyroidism. They are focal bone lesions found in any part of the skeleton. Brown tumors in the craniofacial bones are uncommon. The term "brown tumor" was derived by the color and presence of hemosiderin deposits in the tumor. The appearance of the tumor is also caused by vascularity and hemorrhage. The most common areas of brown tumors include extremities, ribs, clavicles, and pelvic girdle. In patients with craniofacial involvement, there is a significant morbidity. Most patients suffer from social stigma due to facial disfiguration. Patients may also suffer from normal function such as talking, chewing, and breathing. A multidisciplinary team is required for accurate diagnosis, treatment, and management of brown tumors. Herein, we report an unusual parathyroid adenoma presenting as a brown tumor of the mandible.

Key words: Brown tumors, craniofacial involvement, secondary hyperparathyroidism, treatment

INTRODUCTION

Hyperparathyroidism (HPT) is a complex disease attributed with a complex anatomic, clinical, and biochemical abnormalities. HPT results from excess secretion of the parathyroid hormone (PTH). The disease may occur either in tertiary, secondary, or primary forms. Abnormality in one or more parathyroid glands causes an excess in the secretion of PTH.[1] As per evidence, parathyroid adenomas were observed in nearly 85% of primary HPT cases. The appellation "brown tumor" is derived from the color of the tumor present. The appearance was caused by the hemorrhage, vascularity, and deposits of hemosiderin.[2] In cases of primary HPT, bone manifestation is often a late manifestation and accepted by most clinicians.

Parathyroid adenoma is a common cause of primary hypercalcemia. An increased secretion of PTH often causes the typical symptoms of hypercalcemia.

PTH has several receptors that facilitate in raising blood calcium concentrations. PTH acts at the distal renal tubules to reabsorb calcium. PTH also plays a key role in bone resorption by enhancing osteoclastic activity. PTH increases the production of activated Vitamin D by enhancing the absorption of calcium within the intestine.

The term "brown tumor" is used when primary HPT is involved with the bone. This association is also widely known as "osteitis fibrosa cystica." Symptoms of hypercalcemia are common among patients with brown tumors. These tumors are rarely found in parathyroid carcinoma with an exception of few cases.[3]

The appearance of "brown" is associated with the fibroblastic tissue penetration within the gaps of the bone matrix. The tissue penetration is a result of increased osteoclastic activity. One of the major consequences of increased osteoclastic activity includes expansion of the bone beyond the normal contours. In some cases, patients may have bone pain, wherein the disease is involved within the periosteum. As per current evidence, brown tumors have been reported in 4.5% and 1.5% of patients with primary or secondary disease, respectively.[4]

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Brown tumors can be located in any area of the skeleton. However, the most common areas include ribs, extremities, clavicles, and pelvic girdle. Brown tumors within the craniofacial bones are an unusual finding.[5] Herein, we report an unusual case of parathyroid adenoma presenting as a brown tumor of the mandible.

CASE REPORT

A 38-year-old female presented with a large swelling and ulcer over the buccal mucosa in the body of the mandible. The patient reported of pain and discomfort for 1 month. She also complained of mild pain and paresthesia. However, she did not report any weight loss, rigor, or fever. The patient was systematically well with no medical history. The patient had no history of alcohol or tobacco intake.

She was apyretic and systemically well on physical examination. There was no cervical lymphadenopathy. The right cheek was swollen when compared to the contralateral side. However, no evidence of infection was made on clinical assessment. Intraoral examination showed a 3 cm × 4 cm ulceroproliferative growth involving right lower alveolus involving gingivobuccal sulcus. A computed tomography (CT) of the neck and thorax was carried out to evaluate the patient.

A three-dimensional reconstruction of the mandible [Figure 1] revealed a well-defined expansile lytic soft tissue mass lesion involving and arising from the right lower alveolus (molar, premolar, and canine regions) and part of the body of the mandible.

Maximum measurable size of the lesion was 4.5 cm × 3.7 cm × 3.6 cm. The lesion was medially abutting the right lateral border of the tongue and laterally abutting the buccal space.

Screening cervical level shows few nodes in the submental and right IB region.

Contrast CT axial [Figure 2] and coronal [Figure 3] scan showed mild enhancement of above-mentioned lesion. Minimal enhancement of nodes with maintained fatty hila were observed.

Lower cervical region showed enhancing hypodense lesions in the region of the right superior parathyroid and inferior parathyroid each measuring 13 mm × 12 mm and 12 mm × 10 mm, respectively, representing features of parathyroid adenoma.

The surgeon performed an excisional biopsy followed by curettage of the bony walls with a large round bur. Histological slide stained with hematoxylin and eosin showed giant cell bone tumor including multiple giant cells, spindle-shaped stromal cells, fibrous connective tissue proliferation, and areas of hemorrhage characteristic of brown tumor.

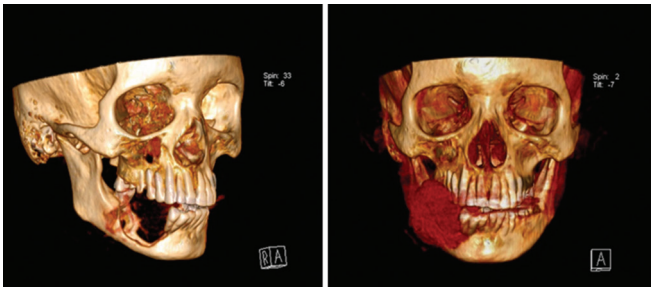

Figure 1: Three-dimensional reconstruction of the mandible

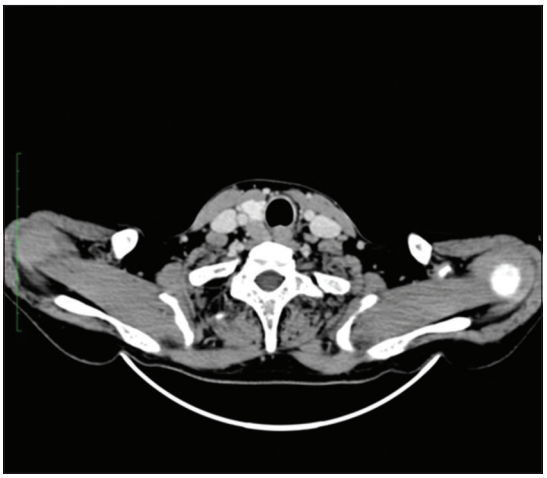

Figure 2: Contrast axial computed tomography scan

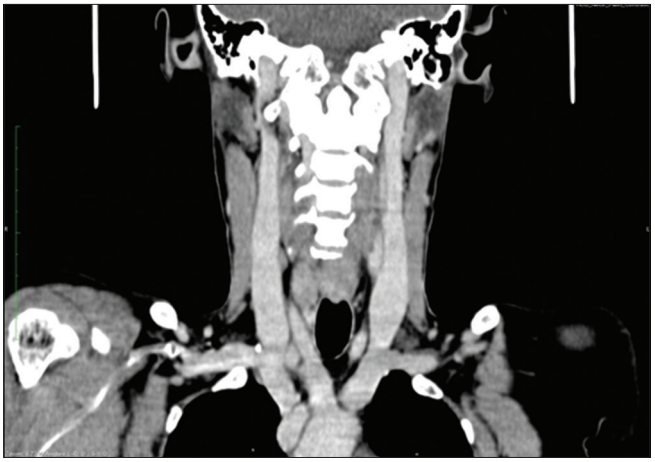

Figure 3: Contrast computed tomography coronal [Figure 2]

Blood biochemistry revealed elevated serum calcium level 13.52 mg/dl, alkaline phosphatase 211 IU/L, parathormone 92 pg/ml, and phosphorus 3.9 mg/dl. The patient underwent surgery for the adenoma. The post-operative event was normal.

DISCUSSION

Brown tumors are non-malignant giant cell lesions that are associated with an increase in circulating PTH levels. Brown tumors are a representation of terminal stages of bone remodeling in the hyperparathyroid state. The incidence of the brown tumor is less uncommon with 1.5–1.7% occurring in secondary HPT while 4.5% occurring in primary HPT. The overall incidence of brown tumors is 0.1%.[6]

HPT can be classified as primary, secondary, or tertiary based on the underlying mechanisms. Primary HPT is caused due to hyperplasia, malignant or benign neoplasm of one or more parathyroid glands. Secondary HPT is caused due to Vitamin D deficiency or hypocalcemia. It may be caused secondary to chronic renal insufficiency. Tertiary HPT is caused due to renal failure. However, another unique type of HPT has been recognized, which is caused by increased PTH levels among patients with malignant disease.[7]

The diagnosis of brown tumors is based on presumption, wherein the differential diagnosis is based solely on histological findings. The definitive diagnosis of brown tumors is made on key radiological and laboratory data. Brown tumors usually form at the base of the skull, paranasal sinuses, orbits, spinal column, humerus, tibia, and clavicles. It is also formed within the jaws of young patients. However, brown tumors in the maxilla are an uncommon finding.[8]

As per clinical presentation, brown tumors are commonly small and asymptomatic (with swelling) in the jaw bones. They may also be painful exophytic mass. Radiologically, the lesions appear well-demarcated multiocular or monocular osteolytic lesion infrequently attributed to loss of lamina dura and root resorption. The diagnosis of brown tumors can be confirmed by examining elevated serum PTH and calcium levels.[9,10] Histological features alone cannot be relied on since they may resemble a giant cell tumor. In our case, radiological, histological, and biochemistry results confirmed the diagnosis of brown tumor.

Based on current evidence, the mandible has a higher rate of being affected as compared to the maxilla. Brown tumors remain fairly asymptomatic with an exception when they are large.[11] In our case, the patient was asymptomatic but reported of pain and discomfort as the tumor got large. Brown tumors are known to cause facial disfiguration which negatively impacts the patient's quality of life and social ease. In most cases, the patient's breathing, chewing, and talking capabilities are affected. Some of the other uncommon complications include proptosis of the eyes, headaches, displacement/mobility of the teeth, visual impairment, and nasal/intraoral bleeding.[11] In our case, the patient did have a social stigma accompanied with pain and discomfort.

CONCLUSION

Brown tumors are rarely observed in recent times as most patients are managed before the onset of primary tumor development. Brown tumors are associated with significant morbidity or key message to clinicians is to keep in mind of this condition when patients present with tissue swelling without symptoms of hypercalcemia. The accurate diagnosis of this condition prevents avoidable local surgery. The preferred choice is to treat the primary endocrine abnormality.

- Patients with a soft tumor of the mandible should be primarily assessed for hypercalcemia
- If the brown tumor is confirmed, the primary treatment should involve parathyroidectomy
- A multidisciplinary approach with the help of endocrinologists, otolaryngologists, radiologist, and otolaryngologists should be considered.

ACKNOWLEDGMENT

We would like to thank Mr. Lyndon Fernandes for his medical writing assistance.

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How to cite this article: Pawar S, Patil A, Borde C, Nagarkar R. An Unusual Presentation of Parathyroid Adenoma: A Radiological Point of View. J Clin Res Radiol 2019;2(1):1-4.